DESMOID TUMORS*

A CLINICAL AND PATHOLOGIC STUDY

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DESMOID TUMORS are benign fibromata arising from musculo-aponeurotic structures. According to MacCarty,[29] they are fibrocytomata occurring in striate muscle. The majority of them occur in the anterior abdominal wall, but they also occur, not infrequently, in other parts of the body.

This study is concerned with the clinical and pathologic manifestations of the 77 desmoid tumors which have been diagnosed at the Mayo Clinic during the past 30 years (January 1, 1908, to December 31, 1937, inclusive). The histories of these cases have been carefully abstracted, with especial reference to sex, age, symptoms, known duration of tumor, trauma, previous operations, pregnancies, treatment and recurrence, where definite follow-up information was available. The gross specimens were studied, and microscopic sections were made of each tumor in order to study cellular detail.

In order to be able to compare the results of this study with the results of those reported in the literature, desmoid tumors have been divided into two groups: (1) Those occurring in the anterior abdominal wall; and (2) those occurring in the musculo-aponeurotic structures elsewhere in the body.

The history of desmoid tumors apparently began in 1832, with the report of two cases by Macfarlene, of Glasgow. Although he did not coin the term "desmoid," there can be little doubt, from the details given, that the tumors were examples of fibroma of the abdominal wall. The first was described under the title, "Organized Sarcomatous Tumour between the Layers of the Abdominal Muscles," and the other as a "Fibrocartilaginous Tumour" in the same situation. The first patient was cured by excision of the tumor; the second patient died of peritonitis following excision of the tumor.

Müller, in 1838, first suggested the use of the term "desmoid" because of the band and tendon-like arrangement in these fibrous tumors. However, the tumors were called, and generally referred to by the French term *"fibrome de la paroi abdominale"* until Sänger (1884) revived the term "desmoid" in conjunction with his operative innovation of resecting the peritoneum in

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cases in which this membrane was adhered to the growth rather than shelling out the tumor from its false capsule as previously had been done.

According to Labbé and Remy, nine cases of desmoid tumors had been reported prior to 1850. The fibrous nature and benign character of these tumors were recognized from the onset, although there was much controversy as to their point of origin. Huguier (1860) correctly stated the fibrous nature of the growth, but believed that it arose by a pedicle from the periosteum of the pelvis, and suggested section of the pedicle as a method of treatment.

While the French were being misled by Huguier, Cornils (1865) and Buntzen (1868) developed the musculo-aponeurotic theory of origin and suggested treatment by excision. Guyon, in 1877, corrected the error initiated by Huguier. In 1875, Suadicani made the great advance of applying listerism to the extirpation of these tumors.

Except for occasional case reports, the term "desmoid" was seldom seen in the literature until 1890, when Ledderhose reviewed 100 cases reported in the literature, and, in 1904, when Pfeiffer, in a monograph, collected and analyzed 400 cases (40 of his own and 360 reported in the literature). Since then various case reports and summaries have appeared in the literature, such as those of Stone, Balfour, Stewart and Mouat, Bessesen, Bevan, Cullen, Delbet, Marlow, Polacco, Powers, Walters and Church, Nichols, Danforth, Cahn, Andrews, Mason, Penick, and Geschickter and Lewis.

Mankin collected, from the literature, reports of 629 cases of neoplasms of the anterior abdominal wall; in 423 of the cases the neoplasm was a fibroma and in 152 it was a fibrosarcoma.

Most of the reports have concerned desmoid tumors of the abdominal wall; however, in a number of cases of desmoid tumors, such as those reported by Bellanger, Cigolini, Salto, Auvray,[2, 3] Hoffmeister, Mason and Esau, the tumors occurred elsewhere in the musculo-aponeurotic structures of the body.

Desmoid tumors are of comparatively infrequent occurrence. Gurlt, of Vienna, found only eight in a study of 16,637 tumors. Labbé and Remy observed ten cases during 20 years; Billroth, 16 in 23 years; Nélaton, 15 in 26 years; Pfeiffer, 40 cases at von Brun's Clinic, at Tubingen, in 46 years, and Mason, 50 cases during 24 years.

Although the real cause of these tumors still remains unknown, much has been written on this phase of the subject.

In the 400 cases included in the monograph by Pfeiffer, 87.1 per cent of the patients were women. Data regarding the occurrence of pregnancy were available in 265 of the cases in which the patients were women. In 250, or 94.3 per cent of the 265 cases, the patients had borne children. In 93 per cent of the cases reported by Stewart and Mouat the patients were women, and 80 per cent of these women had borne one or more children. In most of the cases reported by Stewart and Mouat the tumor occurred in the third or fourth decade of life. Geschickter and Lewis reported 20 cases of desmoid tumors of the abdominal wall. In 19 of the cases the patients were females; in 17 of the 19 cases the patients were between the age of puberty and the

menopause. All of the tumors were related to pregnancy or the scar of an operation. These authors stated: "The rarity of such fibromata . . . is in keeping with the simple and direct formation of adult fibrous tissue from mesenchyme. The simplicity of this differentiation in a tissue which retains its power of regeneration supplies very few 'embryonic rests,' in the sense of Cohnheim, from which tumors might arise." These authors expressed the opinion that there is some relationship between these growths and sex physiology.

Stewart and Mouat said that there was a history of trauma or severe muscular strain in nine of their cases and that the tumor occurred in an operative scar in five cases. Penick, in a review of the literature, found reports of 15 cases in which the tumor arose in operative scars and added two cases in which this occurred. Auvray,[2] Esau, Hoffmeister, and Cigolini also reported trauma as a possible causal factor. Ebner, in 1880, advanced the theory now held by MacCarty,[26] namely, that these tumors result from muscular rupture either in consequence of violent effort or traumatism. Labbé and Remy (1888) suggested that muscular rupture during violent contractions of labor was the real explanation, that is, the physiologic trauma of labor.

In view of the traumatic theory, Nørgaard contrasted the comparatively common hematoma of the rectus muscle caused by severe strains or infectious diseases with the rare spontaneous hematoma in the same muscle. In all of the 72 cases which he reported, the patients were women, and all of the women except one were multiparae. In 11 cases spontaneous hematoma occurred during pregnancy. The clinical picture was usually misinterpreted and practically all of the patients were treated surgically.

It appears highly probable that this condition may be the basis of desmoid tumors, that is, organization in an hematoma and subsequent hyperplasia, the cause of the latter being unknown. Repetto expressed the opinion that a desmoid tumor is the result of an inflammatory reaction in a small hemorrhage in the abdominal wall followed by fibrosis and hyperplasia.

Various clinical observations have been reported in regard to these tumors. Stewart and Mouat said that the tumor is usually discovered accidentally by the patient or by a physician after some slight trauma has led to physical examination. The presence of the tumor may occasionally cause pain of a localized or radiating nature. Even when the tumor has attained considerable size it rarely causes pressure symptoms, and signs and symptoms of intra-abdominal involvement are lacking. The tumor does not involve the skin, which can be moved freely over the tumor. With the abdominal muscles relaxed, the tumor can be moved about in certain directions with sufficient freedom to show that it is unconnected with any intraperitoneal structure. It is also immobilized by contraction of the muscle or muscles in which it lies and recovers its mobility when the muscles are relaxed. It must be considered in the differential diagnosis of tumors occurring in operative scars, as pointed out by Balfour, Danforth and Mason. Cigolini, Salto, Auvray[2, 3] and Esau have pointed out that the tumor must be suspected in all cases in which a

tumor occurs in muscle and that a biopsy should be performed to determine its exact nature.

The gross and microscopic appearance has been well-described by various authors. Stewart and Mouat reported that the tumors always occur singly. They may be oval, round or flat, and they may vary in size from 1 to 12 cm. in diameter. Rokitansky (1880) described one weighing 17 Kg. Montgomery and Bland (1905) reported a case in which the tumor weighed 19 pounds (8.6 Kg.). The tumor may become adherent to the fascia or peritoneum. Owing to the pressure of the growing tumor against adjacent structures it may appear encapsulated. It is densely hard, white or pinkish, and cuts with difficulty.

The microscopic structure, according to Geschickter and Lewis, is that of a rather cellular fibroma occurring in striate muscle. The tumor is clearly an infiltrating type, without a capsule. The especial features of the tumor are the inclusion of striate muscle, as described by Durante (1902).

The treatment, according to most of the writers on the subject (Balfour, Mason, Stewart and Mouat, Pfeiffer, Penick, and Geschickter and Lewis), is surgical.

The prognosis, according to Geschickter and Lewis (1935), is good, as the majority of their patients lived well beyond the five-year period, after surgical removal of the tumor. Pfeiffer (1904) in a postoperative study of 107 cases found a recurrence in 33, or 30.8 per cent. In Stone's first case (1908) the tumor recurred and was again excised, 20 years after the first operation. In Morison and Drummond's case there was no evidence of recurrence two years after the second excision.

ANALYSIS OF CASES

An analysis of the present series of 77 cases of desmoid tumor reveals that in 55 cases the tumor occurred in the anterior abdominal wall, and in 22 cases it occurred elsewhere in the skeletal muscular system (Table I).

TABLE I

SITE OF DESMOID TUMORS

Site of Tumor	No. of Cases
Anterior abdominal wall (rectus abdominis, external or internal oblique, or transversalis muscles)	55
Pectoralis major	5
Muscles about the scapula	4
Rectus femoris	3
Gluteal muscles	2
Sternomastoid muscle	1
Posterior belly of digastric muscle	1
Biceps brachii	1
Extensor carpi ulnaris	1
Hamstring muscles (biceps femoris, semitendinosus and semimembranosus)	1
Dorsum of foot	1
Masseter	1
Scar of radical operation on the breast	1
Total	77

The following is an analysis of the 55 cases in which the tumors occurred in the anterior abdominal wall. In 40 cases, the patients were females, and in 15 cases, the patients were males. Most of the tumors occurred between the ages of 20 and 40, among the females, and between 40 and 60, among the males. The size of the tumors varied widely; in most cases the tumors were between 3 and 8 cm. in diameter. The known duration of the tumors varied from one month to several years. No relationship was noted between the size of the tumor and its known duration. There was a history of pregnancy in 25 of the 40 cases in which the patients were females. A history of trauma was obtained in four cases, and in 14 cases the desmoid tumor occurred in an operative scar. In 47 cases the patients were treated by excision, and in eight cases they were treated by a combination of excision and irradiation. Follow-up information was available in 47 of the cases in which the tumor occurred in the anterior abdominal wall. In 39 of the 47 cases excision only was employed, and in the remaining eight cases excision and subsequent irradiation were employed. Local recurrence of the tumor was noted in three cases in which the treatment consisted only of excision of the tumor, and in one case in which irradiation was employed following excision. In two of the three cases in which a local recurrence followed treatment by excision only, the recurrent tumors were excised, and follow-up data are available for eight and ten years, respectively, after the second operation. The tumor did not recur after the second operation in either case. In the 55 cases in which the tumors involved the anterior abdominal wall, 20 patients were known to be alive ten years, and 16 patients are known to be alive five years, respectively, following the completion of treatment. There were 20 known ten-year cures and 16 known five-year cures in this group of cases.

The following is an analysis of the 22 cases in which the tumor occurred elsewhere in the skeletal muscular system than in the anterior abdominal wall. The site of the tumors is shown in Table I. In the 22 cases, seven patients were males and 15 were females. Most of the tumors occurred between the ages of 20 and 40, in the females, and 40 and 60, in the males. The size of the tumors varied from 1 cm. to 10 cm. in diameter. The known duration of the tumors varied from one month to 24 years. No relationship was noted between the size and known duration of the tumor. There was a history of pregnancy in eight cases, and a history of trauma was obtained in two cases. The tumor occurred in an old operative scar in only one case (Table II). In 16 cases, the patients were treated by excision only, and in six cases, the patients were treated by excision combined with irradiation therapy. Follow-up information was available in 15 cases. In ten of the 15 cases, treatment consisted of excision only; in the remaining five cases, excision was followed by irradiation. There was a local recurrence of the tumor in the ten cases in which excision only was employed, but no local recurrence was observed in the five cases in which excision was followed by irradiation. In the 22 cases, in which the tumors were situated elsewhere than in the anterior abdominal wall, three of the patients have lived for ten years, and four patients

have lived for five years, respectively, following the completion of treatment (Table II).

TABLE II

CLINICAL DATA AND RESULTS OF TREATMENT IN 77 CASES OF DESMOID TUMOR

	Sex					Treatment		Results					
								Patients Known to have Lived 10 Years Following Treatment		Patients Known to have Lived 5 Years Following Treatment		Recurrence of Tumor	
Site of Tumor	Males	Females	History of Pregnancy	History of Trauma	Tumors Occurred in Operative Scar	Excision Only	Excision and Irradiation	In Cases in which Excision Only was Employed	In Cases in which Excision and Irradiation were Employed	In Cases in which Excision Only was Employed	In Cases in which Excision and Irradiation were Employed	In Cases in which Excision Only was Employed	In Cases in which Excision and Irradiation were Employed
Anterior abdominal wall	15	40	25	4	14	47	8	18	2	12	4	3*	1
Other parts of body	7	15	8	2	1	16	6	1	2	2	2	3	
Total	22	55	33	6	15	63	14	19	4	14	6	6	1

* In two of these cases the recurrent tumor was excised; in the two cases there was no evidence of a second recurrence eight and 12 years, respectively, after the second excision.

The following is an analysis of the 77 cases. Twenty-two of the patients were males and 55 were females. A history of pregnancy was obtained in 33 cases, a history of trauma was obtained in six cases, and the tumor occurred in an operative scar in 15 cases. In 63 cases, treatment consisted of excision only; in the remaining 14 cases, excision was followed by irradiation. There was a local recurrence of the tumor in six, or 9.5 per cent, of the 63 cases in which excision only was employed, and in one, or 7.1 per cent, of the 14 cases in which excision was followed by irradiation. There were 23 known ten-year cures and 20 known five-year cures.

There was only one postoperative death in the entire series of 77 cases. This was due to infection of the operative incision following the removal of a large desmoid tumor from the anterior abdominal wall (an operative mortality of 1.3 per cent).

CLINICAL ASPECTS

Desmoid tumors must be considered in the differential diagnosis of all masses that are situated in or come in contact with the musculo-aponeurotic structures of the body. In this series of cases the patients, generally, sought aid because of the presence of a tumor mass. Other less frequent complaints were: A dragging sensation or sense of weight in the abdomen; a mild sticking pain; a sudden increase in size of a previously small nodule; a bizarre pain in the abdomen; and a sense of pressure on the bladder.

In this series of cases the desmoid tumors were found more frequently in the anterior abdominal wall than elsewhere in the body and, occasionally, had

been diagnosed by the clinician as being intra-abdominal. They had been mistaken for hydrops of the gallbladder; a large stone in the gallbladder; a tumor of the omentum or mesentery; pancreatic cyst; a uterine tumor, a tumor of bone; and a tumor of kidney. When situated elsewhere in the body they generally had been mistaken for sarcomata.

The following points have proved of value in the clinical diagnosis of these tumors. As a rule the tumor is smooth, round or flat, firm and discrete. The tumor is situated below the skin but is not attached to it. The tumor is movable, moves with the muscle to which it is attached, and is immobilized by contraction of this muscle. On deep inspiration the tumor moves forward but not downward as do most intra-abdominal tumors.

A desmoid tumor must be distinguished from keloids, fibrosarcomata, inflammatory conditions, and lesions which extend secondarily to the abdominal wall from nearby organs. A keloid will, more frequently, be difficult to distinguish from a desmoid tumor than will other conditions mentioned because this overgrowth of fibrous tissue is closely allied to the desmoid tumor and resembles it in many respects. The common keloid involves the skin and subcutaneous tissues to form raised regions covered with shiny epithelium; in this respect it differs from a desmoid tumor, which involves the skin occasionally, and then only secondarily, and is usually situated below the skin. These two conditions can also be differentiated microscopically. The keloid is relatively acellular with conspicuous tracts of collagen; the remains of obliterated capillaries are usually visible. A desmoid tumor is more vascular and more cellular than a keloid and evidences of cellular activity are more noticeable.

According to MacCarty,[27, 28] the only way to be certain of the diagnosis is to remove a specimen at the time of operation and examine it microscopically.

GROSS AND MICROSCOPIC PATHOLOGY

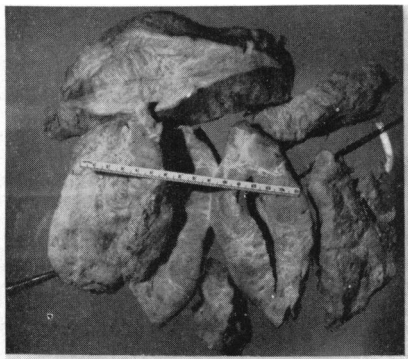
A freshly removed tumor is dense, hard and tough, and creaks under the knife. The cut surface bulges, is white or pinkish in color, glistens, and the interlacing bundles of white fibrous tissue are usually seen without difficulty (Fig. 1). The larger tumors tend to be soft in the center, either from edema or mucoid degeneration, and some are cystic. Changes in the skin covering the tumor are unusual. There was no associated involvement of the lymph nodes in this series of cases.

The histologic picture is that of a rather cellular fibroma occurring in striate muscle. The central portion of the tumor proved to be older than the peripheral portion. The elongated adult fibrous tissue cells run in strands and bundles which are interlaced in all directions after the manner of the unstriate muscle bundles in a uterine fibroid (Figs. 2 and 3).

At the periphery, where the tumor infiltrates the surrounding muscle, there is a tendency for the tumor to be somewhat more cellular. There is no capsule or definite line of cleavage between the tumor and the adjacent muscle. It is only when the tumor is bounded by a fascial plane that the appearance of

encapsulation is produced (Fig. 4). In spite of this infiltration there was no evidence of a sarcomatous change, such as nuclei containing large amounts of darkly-stained chromatin or large multiple nucleoli, both of which are indications of rapid proliferation. Normal adult blood vessels were noted throughout most of the tumors. These vessels were an indication of the slow growth of the tumor.

FIG. 1.



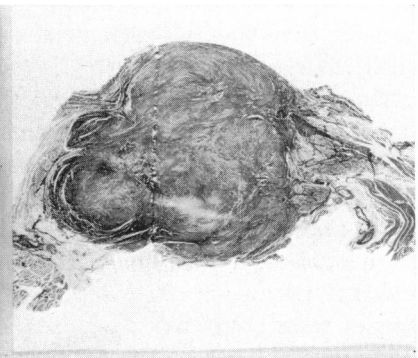
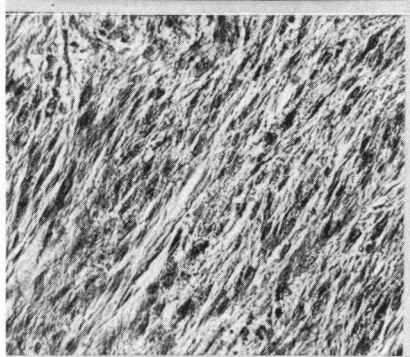


FIG. 4.

FIG. 2.



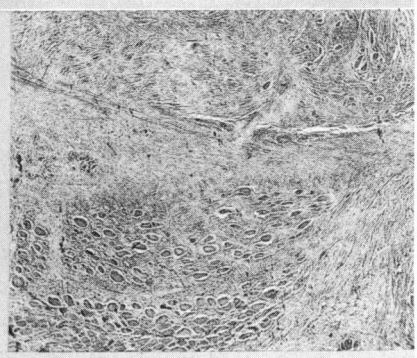
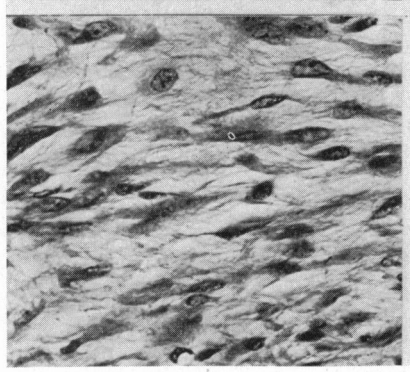


FIG. 5.

FIG. 3.



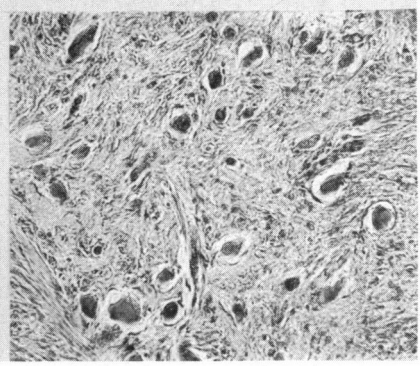


FIG. 6.

FIG. 1.—Desmoid tumor removed from abdominal wall, showing the characteristic band-like arrangement.
FIG. 2.—Cellular character of desmoid tumor. (×285)
FIG. 3.—Cellular but benign appearance of a desmoid tumor. (×450)
FIG. 4.—Cross-section of tumor lying between separated fibers of the rectus abdominis muscle in the rectus sheath; the tumor appears to be encapsulated. (×1¾)
FIG. 5.—Infiltration of muscle fibers by a desmoid tumor. (×30)
FIG. 6.—Muscle with "foreign body-like" giant cells. (×195)

The special features of the tumor were the inclusion of striate muscle fibers and the sequence of regressive changes. The earliest change was seen at the periphery of the tumor where the striate muscle was being infiltrated and broken up into constituent fibers (Fig. 5).

The muscle fibers appeared to be stretched and attenuated. They were irregular in outline and the transverse striations tended to disappear. Here and there were strands resembling unstriate muscle and "foreign body-like" giant cells derived from this muscle probably because of faulty nutrition (Fig. 6).

It is easy to see that failure on the part of the surgeon to remove the tumor completely would leave strands of interfibrillar desmoid tissue and result in a local recurrence of the tumor.

COMMENT.—The cause of these tumors as yet remains unknown. However, the most logical theory of origin seems to be one of trauma superimposed on individual predisposition. A history of trauma was obtained in 21 cases (accidental trauma in six cases and operative trauma in 15 cases), and a history of pregnancy (physiologic trauma of labor) was obtained in 25 cases. Thus trauma appeared to be an etiologic factor in 46, or 60 per cent, of the cases studied. The endocrine theory, as related to pregnancy, seems void because 22 patients were males and only 33 of the 55 females had been pregnant.

No differences in etiology, clinical findings, pathology, curative treatment, or prognosis could be noted between the tumors which occurred in the anterior abdominal wall and those which occurred in other parts of the body.

At first glance, it would appear that a combination of excision and roentgenotherapy would be the ideal treatment, since there was a recurrence in 9.5 per cent of the cases in which the patients were treated by excision only, and in only one, or 7.1 per cent, of the cases in which the patients were treated by a combination of roentgenotherapy and excision. However, several factors have to be considered: (1) The small number of cases in which the patients were treated by excision and radiation (14) as compared to the number treated by excision only (63), (Table II); (2) the insignificance of the difference in number of five-year and ten-year cures obtained by the two methods (Table II); (3) no definite criteria were used for the employment of roentgenotherapy—it was employed in some cases in which the tumor was completely removed surgically but was not used in some cases in which the tumor was large or difficult to remove; and (4) a few of the patients had been treated unsuccessfully by roentgenotherapy only before they finally were cured by surgical treatment.

SUMMARY

This paper is based on a study of 77 cases of desmoid tumors. In 55 cases the tumor occurred in the striate muscle of the anterior abdominal wall, and in the other 22 cases it occurred in striate muscle elsewhere in the body. A desmoid tumor is a simple fibrous tumor that arises in musculo-aponeurotic

structures and tends to infiltrate the muscle in which it lies. Trauma (accidental, operative, or the physiologic trauma of labor) combined with an unknown individual predisposing factor, appears to be the most logical theory of origin. There was a peculiar change in the striate muscle fibers enclosed in the growth. This change appears to be a process of differentiation and results in the formation of multinucleated plasmodial masses resembling foreign body giant cells. These tumors do not undergo metastasis nor do they endanger life; however, they do tend to recur locally, unless completely removed. Diagnosis is made by finding a fibrous tumor in muscle; the tumor is fixed by contraction of that muscle. A biopsy is indispensable for diagnosis. Treatment consists of complete excision. Roentgenotherapy appears to be of little, if any, additional value. The operative mortality in this series of cases was 1.3 per cent. There was a local recurrence of the tumor in seven, or 11.8 per cent, of the 69 cases in which follow-up data were available. We did not discover any evidence that this tumor ever undergoes sarcomatous change.

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